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Duplicate pancreas meets gastric duplication cyst: A tale of two anomalies

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ABSTRACT

INTRODUCTION: Congenital anomalies are a rare cause of pancreatitis in adults. Gastric duplications are the least common duplication of the gastrointestinal tract and are even more uncommon in the setting of a duplicate pancreas.
PRESENTATION OF CASE: This manuscript contains a case report and review of the literature of an adult who presented with recurrent pancreatitis and was found to have a gastric duplication cyst that communicated with a duplicate pancreas. The study aim is to alert practitioners to the duplicate anomaly and recommend appropriate therapy.
DISCUSSION: Combined gastric and pancreatic duplications usually occur in young females with nonspecific, recurrent abdominal pain. This combined duplication can result in pancreatitis when the gastric duplication is contiguous with the stomach. Heightened awareness of the condition, appropriate diagnostics with accurate interpretation and a minimalist approach to resection are warranted.
CONCLUSION: Recurrent abdominal pain and pancreatitis in young adults devoid of risk factors should lead to consideration of congenital anomalies. Not all cysts near the pancreas and stomach are pseudocysts. ECRP and abdominal CT/MRI provide critical diagnostic information. This dual anomaly is best treated by simple excision of the gastric duplication and heterotopic pancreas.

1. Introduction

Developmental anomalies are a rare cause of pancreatitis in adults. Gastric duplications are congenital anomalies that result from abnormal foregut development and are the rarest of all duplications of the gastrointestinal tract.[1–3] This is a case report and literature review of an adult presenting with recurrent pancreatitis that was found to have a contiguous gastric duplication cyst that communicated with the main pancreatic duct through the tail of a duplicate pancreas. The aim of the study is to alert practitioners to this duplicate anomaly and recommend appropriate diagnostics and treatment based on review of the literature.

2. Presentation of case

This 43 year old Caucasian male product of a twin birth presented with recurrent episodes of brief, nonradiating, epigastric and right upper quadrant abdominal pain since age 14. Over time, the pain escalated in frequency and duration lasting up to several days. Prior to referral he was hospitalized with a diagnosis of pancreatitis. The patient was a nondrinker, had never been jaundiced and was not diabetic. Past medical history included hypertension, a non-sustained episode of SVT and elevated lipids. Lipids normalized with minor dietary changes. Liver function tests and Ca 19-9 were normal. Amylase and lipase were elevated during episodes of pancreatitis, but then normalized.

An abdominal computed tomography (CT) scan revealed a heterotopic pancreas with "pseudocysts" and an extrinsic mass compressing the antrum (Fig. 1a and b). An upper gastrointestinal series (UGI) showed a nonobstructive pattern (<1 h transit time to the colon), minimal irregularity of the duodenal bulb and no evidence of mass compression (Fig. 2). ERCP revealed two separate pancreatic ducts, the first situated in the usual location and the second, originating from the mid to distal body of the pancreas. The second pancreatic duct looped back across the midline and filled a space consistent with a small pseudocyst (Fig. 3). Esophagogastroduodenoscopy revealed extrinsic compression of the distal ventral stomach and mild gastritis. Treatment with proton pump inhibitors and subsequent laparoscopic cholecystectomy failed to relieve the patient's symptoms.

The patient was referred to gastroenterology for "pseudocyst" drainage. However, after multidisciplinary review, the problem was felt to be related to the duplicate pancreas. It was known that a duplicate pancreas could be associated with duodenal or gastric duplication cysts but this was not appreciated on the patient's preoperative imaging.

The patient was explored. The duplicate pancreas was the size and appearance of a normal pancreas, but was heterotopically

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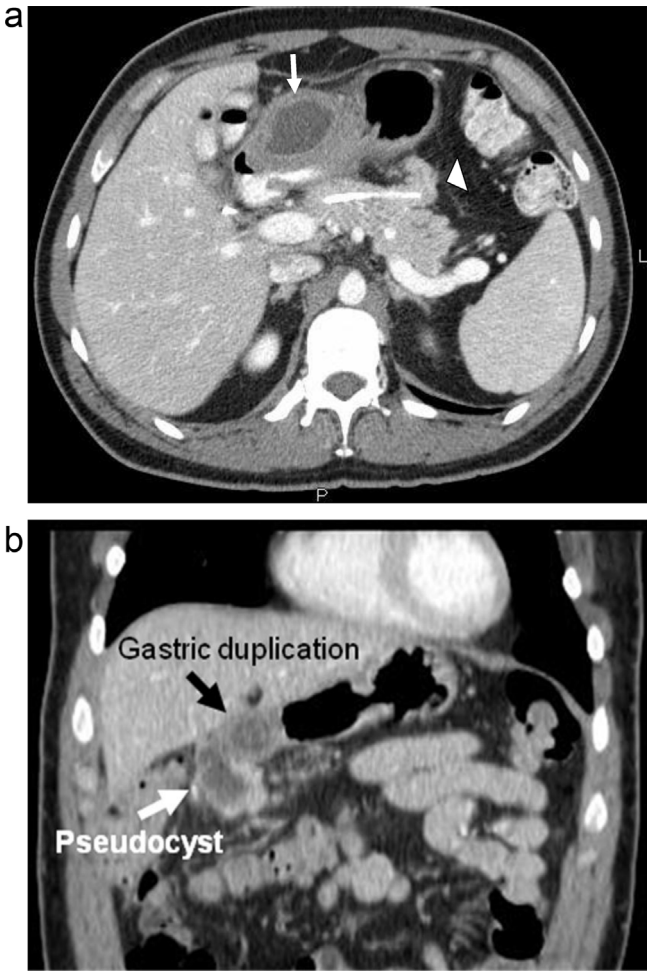

Fig. 1. (a) Axial CT imaging of the abdomen illustrating a gastric duplication within the antrum of the stomach (arrow) and a duplicate pancreas with an indwelling pancreatic duct stent (arrow head). (b) Coronal CT imaging of the abdomen illustrating two cystic structures; one structure emanated from the antrum of the stomach and proved to be a gastric duplication and the other was a pancreatic pseudocyst that arose from the duplicate pancreas (not seen).

located (Fig. 4). The duplicate pancreas emanated from the distal body of the normal pancreas, looped back to the right of midline, and ended in an inflammatory cyst adjacent to the stomach antrum. This cyst cavity was aspirated and filled with amylase-rich fluid. The duplicate pancreas was divided from the main pancreas and resected to the level of the cyst. The cyst was then resected, sent for

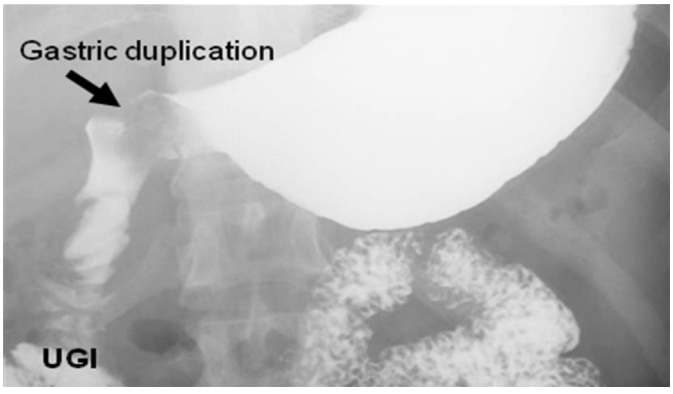

Fig. 2. Upper GI series showing contrast waste within the distal antrum which proved to be the location of the gastric duplication.

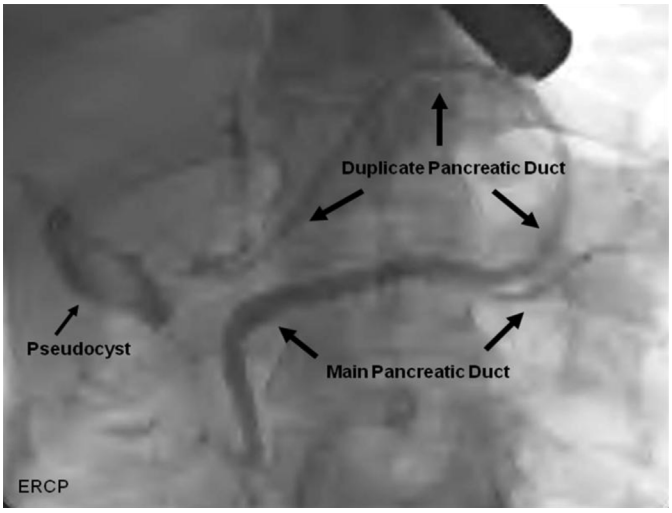

Fig. 3. ERCP image that shows filling of the pancreatic duct of a normal pancreas as well as of the duplicate pancreas. The duct of the duplicate pancreas arises from the mid body of the main pancreas and loops back to end in a cystic structure which proved to be a pancreatic pseudocyst.

frozen section and histologically proven to be a pseudocyst. A fistula tract proceeded cephalad from the pseudocyst into the region of the gastric antrum. By ultrasound, a second thick-walled cystic structure was identified. A catheter was placed in this fistula tract and a sinogram performed. A cystic cavity filled from which no exit was seen. The second cyst was locally resected from the gastric antrum without antrectomy. There was no blood, nor ulcers seen within this second cyst. Pathology confirmed a duplicate pancreas complete with its own pancreatic duct communicating to a pancreatic pseudocyst and contiguous gastric duplication cyst (Fig. 5). The patient had an uneventful post-operative recovery and is living a pain-free, normal life.

3. Discussion

Congenital anomalies are rarely considered in the differential diagnosis of adult pancreatitis. Gastric duplications account for 3.8% of all duplications of the gastrointestinal tract[1,2] occurring less

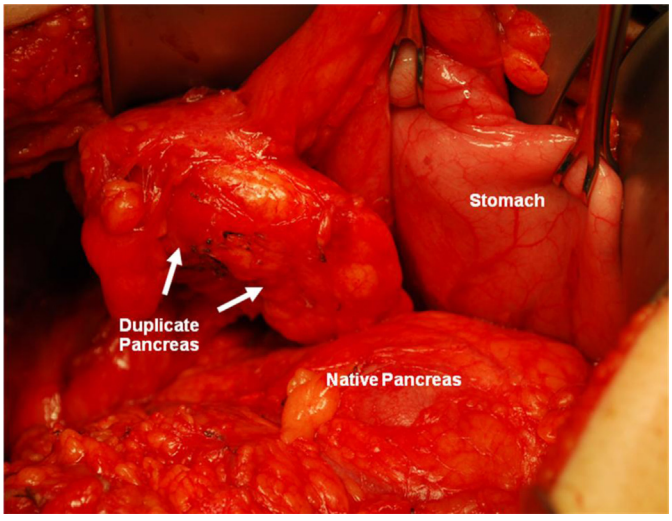

Fig. 4. Intraoperative photo with the lesser sac widely opened and the stomach elevated by babcock clamps; There is a normal pancreas situated in the usual anatomic location and a second complete pancreas emanating from the body of the normal pancreas whose tail is in continuity with the antrum of the stomach.

Table 1
Reported cases of combined gastric and pancreatic duplications.

Reference	Age	Sex	History	Pathology	# ops	Final operation	Contiguous with stomach	PD
Bradbeer[16]	17	M	6 years abdominal pain/emesis	Gastric cyst laid on pancreatic head communicated with PD	1	Excise gastric duplication; externally drain pancreas	N	Y
Schuster Case records of MGH 1964[23]	5	M	3 mo abdominal pain; elevated amylase	Gastric cyst within the pancreatic head, communicated via narrow pedicle of pancreas containing a duct at juncture of body/tail	3	Excise gastric duplication and aberrant pancreas	Y	Y
Katz[17]	34	F	2 years abdo pain, N/V, elevated amylase	Gastric duplications attached to antrum and duodenum and lobular extension of the PD with multiple lumina	1	Excised neck of cyst, pyloroplasty, vagotomy; left pancreas alone	Y	Y
Akers[18]	7	M	18 mo abdo pain; elevated amylase	Gastric cyst within head of pancreas, obliterated ductal connection; pancreatic ascites	2	Excise gastric duplication; pancreaticojejunostomy	N	Y
Akers[18]	21 mo	F	1 year poor feeding; anemia	Gastric cyst in the body/tail communicating with duct of Wirsung; bloody ascites	2	Excise gastric drainage; Roux-Y drainage of pancreas	N	Y
Longmire[11]	15	F	3 years abdominal pain; UGI bleed	2 heterotopic pancreases, originated from neck of normal pancreas and communicate with gastric cyst; massive intraductal hemorrhage/pancreatitis	2	Excise gastric duplication; whipple	Y	Y
Parker[19]	6 mo	F	5 days of BPR, tender mass LUQ	Gastric duplication attached to tail of bifid pancreas and attached to TV colon ulcer/fistula	3	Gastric duplication, adherent TV colon and pancreas tail excised en bloc; partial malrotation noted	N	?
Parker[19]	5 mo	F	LUQ mass	Gastric duplication arose from tail of pancreas	1	Resected gastric duplication and attached normal pancreas	N	?
Traverso[8]	32	F	9 years abdominal pain; elevated amylase	Antral gastric duplication; aberrant pancreas from body of normal pancreas to duplication inferiorly	2	Distal pancreatectomy	Y	N
Rosenlund[20]	31/2	M	4 months of cyclic abdominal pain	Antral gastric duplication–noncommunicating; fibrous cord of pancreas connects with mid pancreas, not PD	1	Excised gastric duplication, excised fibrous cord	Y	N
Schwartz[21]	9 day old	M	Shortly after birth-high fever, FTT, UTI, coffee ground emesis, pneumoperitoneum	Gastric duplication "grew out of the pancreas", malrotation and Ladds bands, colon perf	1	Ladd's procedure, oversewed colon, shelled out of pancreas	N	N
Black[22]	10	F	6 years abdo pain, N/V; amy 352	Gastric duplication adherent to stomach and pancreatic head, fluid amy 640	1	Mass marsupialized into stomach	Y	?
Black[22]	12	F	10 years intermittent pain, N/V	Mass arose from pancreatic head; gastric mucosa, focus of SB; islands of pancreas in muscular wall	2	Resected mass, distal stomach and 1st portion duodenum	?	?
Black[22]	9 mo	M	Emesis after meals, wt loss, dehydration	Gastric dup antimesenteric side of pylorus, attached to pancreas head and PD via small duct	1	Resected anterior part of cyst, oversewed duct, excised mucosa	?	Y
Black[22]	3	M	1 year intermittent abdo pain, N/V	Gastric duplication attached to pancreatic head, small duct communicated with PD, amylase 540	1	Dome of cyst removed, duct oversewn, removed mucosa	?	Y
Spence[5]	8 mo	F	Painful rectal bleeding; anorexia, emesis	Gastric duplication attached to greater curve; attached 6 cm extra pancreas complete with duct	1	Resected duplication and extra pancreas and resected involved colon (ulcer eroded into)	Y	? (Y)
Hoffman[13]	19	F	8 months abdominal pain; elevated amylase	Accessory pancreas with a duct emanating from main pancreas communicated with antral gastric cyst and main PD	1	Excised gastric duplication and aberrant pancreas	Y	Y
Lavine[24]	6	F	2 years abdominal pain; elevated amylase	Gastric antral duplication connected to superior aspect of pancreatic head	1	Excised gastric duplication and aberrant pancreas	Y	Y
Johnstone[25]	41	F	12 years of abdominal pain	Gastric duplication within the pancreatic head/duodenum communicated with PD	4	Biopsy of gastric duplication; Roux-Y drainage of duplication	N	Y
Moss[14]	9	F	5 years abdominal pain	Gastric duplication communicating with aberrant pancreatic lobe arising from pancreatic neck	2	Excise gastric duplication; Roux Y drainage of pancreas	Y	Y
Whidden[26]	24	F	4 years abdominal pain	Aberrant pancreas looped around duodenum communicated with pseudocyst and gastric cyst	2	Excise gastric duplication; remove aberrant pancreas	Y	Y
Muraoka[1]	46	F	10 years abdominal pain, N/V	Aberrant pancreas arising from body of pancreas, connected to gastric cyst that opened into stomach but not PD	1	Excise cyst via gastrectomy; remove aberrant pancreas	Y	N
Christians	43	M	29 years abdominal pain; recent pancreatitis	Aberrant pancreas arising from body of pancreas connected to both pseudocyst and gastric cyst	1	Excise cyst; remove aberrant pancreas	Y	Y

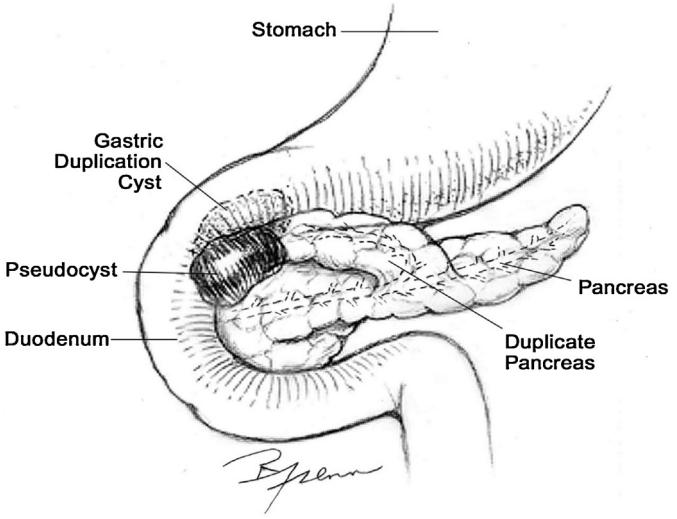

Fig. 5. Line drawing of the dual congenital anomaly illustrating the positions of the duplicate pancreas along with its pseudocyst and contiguous gastric duplication.

commonly than colon, esophageal and ileal duplications.[3] They are even more rare in the setting of a duplicate pancreas. In fact, this is the 23rd reported case of pancreatitis related to the congenital anomaly of a communicating duplicate pancreas and contiguous gastric duplication cyst.

Gastric duplications are cystic structures with (1) an inner alimentary epithelial lining, (2) an outer smooth muscle coat and (3) a blood supply originating from a gastric vessel.[4–6] The duplication may or may not be contiguous with the stomach.[5] Sixty-six percent of gastric duplications occur in the greater curvature, the next most frequent position being the posterior wall (14%).[7] Gastric duplications communicating with the pancreatic duct may adhere to the stomach or may exist as anatomically distinct "cysts". Gastric duplications are almost twice as common in females, and at least one in five have a concurrent intestinal duplication.[8] Gastric duplication cysts result from abnormal foregut development. The two most widely held postulates to explain gastrointestinal duplications are Bremer's errors of recanalization and McLetchie's neuroenteric band theory.[9,10]

Gastric duplications cause symptoms by obstruction of the alimentary tract by regional external pressure, distention of the cystic structure or hemorrhage due to interference with the intestinal blood supply, leading to intestinal mucosal slough.[6] Secretion of gastric enzymes and hydrochloric acid into the confined space of the duplication leads to ulceration and perforation into adjacent viscera with subsequent hemorrhage.

Traverso and Longmire[8,11] distinguished between cases based on contiguity of the gastric duplication to the stomach. They noted that when the gastric duplication cyst was contiguous with the stomach, patients had recurrent pancreatitis. Pancreatitis occurred either due to bleeding into the pancreatic duct (hemoductal pancreatitis) from parietal cells in the duplication causing peptic ulceration[11] or mucoid secretions from the aberrant gastric mucosa entering the pancreatic duct retarding flow and causing obstruction.[8] Recurrent abdominal pain results from pancreatitis or over distention of the gastric duplication and the muscular contractions of the cyst itself.[8,12]

The majority of combined gastric and pancreatic duplication have been found in young females ($n = 15/23$; 69%; median 9 years [9 days–46 years]; 34.8% >16 years). All patients presented with recurrent abdominal pain. Sixty-five percent had contiguous gastric duplications and 78.9% of the gastric duplications were connected to the main pancreatic duct. Ten patients (43.5%) had more than one operation (range 1–4) before correcting the problem and one even underwent pancreaticoduodenectomy. Recently, the majority of patients have had the gastric duplication and the heterotopic pancreas resected with complete symptom resolution.[1,5,8,11,13,14,16–26] (Table 1)

Pancreatitis in younger patients devoid of risk factors warrants further investigation. Diagnostic accuracy is imperative to avoid unnecessary operations and years of morbidity. Our patient's pain started at age 14 and continued for 29 years before the correct diagnosis was made. Axial abdominal imaging, along with interrogation of the pancreaticobiliary ductal system was essential to establish the diagnosis. The unique ERCP finding of an aberrant pancreatic duct terminating in a cyst in a young person with pancreatitis should alert the clinician to this rare, but curable cause of recurrent acute pancreatitis.[13]

In addition to ordering the appropriate diagnostics, it is important to accurately interpret their data. Not all cystic masses near the stomach and pancreas are pancreatic pseudocysts. Our patient had two distinct cystic structures on CT imaging (one being the gastric duplication cyst) but this was not appreciated preoperatively and was instead interpreted as a pancreatic pseudocyst/resolving pancreatitis. Endoscopic ultrasound would likely have shown that one of the two "cysts" was actually within the stomach wall. However, aspiration of cyst fluid may not be definitive as intestinal duplications can contain amylase-rich fluid due to either communication with the pancreas or because of ectopic pancreatic tissue within the duplication itself (10% of duplications).[7,14] In our case, we utilized intraoperative ultrasound (IOPU/S) as per our routine in hepato-pancreato-biliary cases to identify another structure beyond the tail of the duplicate pancreas that was actually embedded within the stomach. Both the finding of the fistulous tract emanating from the duplicate pancreas pseudocyst as well as the IOPU/S allowed us to make the correct diagnosis and render the appropriate surgical therapy.

The optimal treatment of a contiguous gastric duplication that communicates with a duplicate pancreas is local excision of the gastric duplication and duplicate pancreas as well as closure of the pancreatic duct.[1] Because most gastric duplications do not communicate with the gastric lumen, a limited resection is possible (dissect through the muscular wall between the duplication and stomach without entering the lumen) and primarily close the stomach. Marsupialization is inadequate and frequently leads to reoperation.[5] The gastric duplication should be excised not only for symptoms but also because of the risk for developing adenocarcinoma within the duplication.[3,15]

4. Conclusion

Recurrent pancreatitis in young adults without risk factors should lead to consideration of congenital anomalies. Not all cysts near the pancreas and stomach are pseudocysts. ERCP and abdominal imaging provide critical diagnostic information. This dual anomaly is best treated by simple excision of the gastric duplication and heterotopic pancreas.

Conflict of interest statement

None.

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None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

Collection of data was done by Kathleen Christians and Edward Quebbeman. Kathleen Christians wrote the manuscript. They also edited and submitted the final version of the manuscript. Edition of manuscript was done by Sam Pappas, Charles Pilgrim, Susan Tsai and Edward Quebbeman.

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